

Original Article

Gastric diverticulum coexistence with xanthelasma misdiagnosed as a left adrenal gland area cyst: a case report and literature review

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Abstract: The aim of this study was to analyze the clinical data, especially imaging data of gastric diverticulum, which was postoperative pathologically diagnosed as xanthelasma, misdiagnosed as a left adrenal gland area cyst, and review relevant literatures. A gastric diverticulum with xanthelasma case, misdiagnosed as a left adrenal gland area cyst was retrospectively reviewed. Relevant literatures were reviewed to analyze the reasons for misdiagnosis, surgical treatment and pre-operative precautions were summarized. Retroperitoneal laparoscopic left adrenal gland area cystectomy was performed and left adrenal gland was exposed during surgery, but with no detection of the cyst. After Computed tomography (CT) images were reviewed, continual exposure to the abdominal side was conducted until a cyst protruding into the left upper kidney area was observed. Gastric diverticulum was considered after consultation with gastrointestinal surgeons during the surgery, and the diagnosis was confirmed by endoscopic examination and was pathologically confirmed as xanthelasma. It's extremely rare to find gastric diverticulum manifesting as adrenal cyst and it was pathologically confirmed as xanthelasma. Three dimensional CT and dynamic CT examinations were required before surgery and could be used to make differential diagnosis of the left retroperitoneal occupying lesions, particularly in the case adjacent to the gastric area.

Keywords: Gastric diverticulum, gastric xanthelasma, adrenal gland cyst

Introduction

Gastric diverticulum (GD) is a rare disease with low incidence of 0.04 and 0.02% in upper gastrointestinal and autopsy studies, respectively [1]. Much of our knowledge pertaining to gastric diverticula is derived from case reports [2, 3]. It's especially rare to found gastric diverticulum at the left retroperitoneal area [4], Computed tomography (CT) imaging may detect the presence of GDs as thin-walled cystic masses in the left adrenal area. However, sole reliance on CT imaging has been demonstrated to lead to the misdiagnosis of gastric diverticula as adrenal tumors [5, 6]. There is no report on GD diagnosed as xanthelasma by postoperative pathological test and there is no standard for its diagnosis and treatment yet. Therefore, we report a GD postoperative pathologically diagnosed as xanthelasma case that was misdiagnosed as left adrenal gland area cyst, in our hospital and

review relevant literatures to provide diagnosis experience and future guidance for clinicians.

Case description

A 37-year-old female patient was referred to our hospital with chief complaints of shortness of breath for more than 20 days after activities and for further assessment of a left adrenal mass detected by CT. The ultrasound test from the previous hospital reported an anechoic mass (size: 6.8 cm × 5.7 cm × 4.4 cm) located behind the pancreas in the left upper abdomen, being left anterior to the abdominal aorta and anterosuperior to the left kidney, with a good acoustical permeability and a long strip shape boundary distal to the ultrasound transducer. After admission, the recheck of ultrasound test showed an anechoic mass (size: 4.8 cm × 4.7 cm × 4.0 cm) located behind the pancreas in the left upper abdomen, and at the left side of

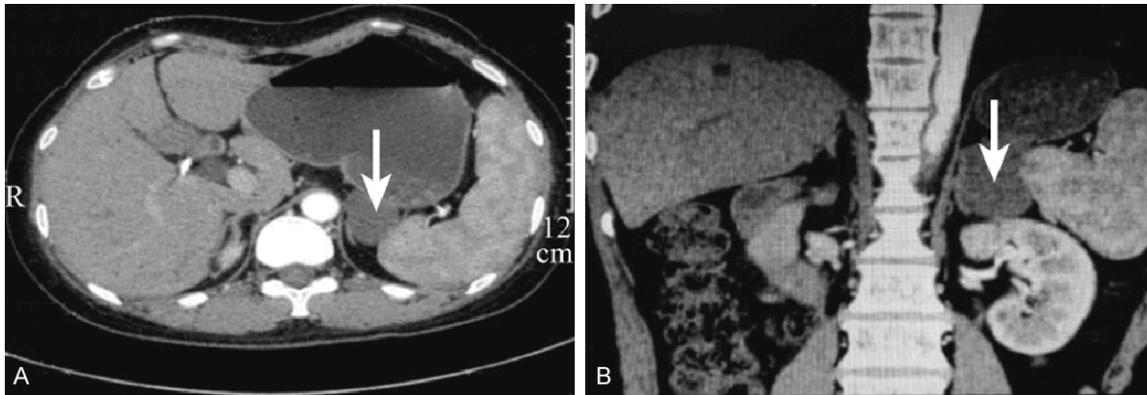


Figure 1. The axial (A) and coronal reconstruction (B) contrast enhanced CT images show that a cystic lesion with size of 48 mm × 47 mm × 40 mm, located in the left side of adrenal region (arrow), is clearly demarcated from adjacent structures.

the abdominal aorta, with clear boundary and relatively regular shape. Color Doppler Flow Imaging (CDFI) test showed no obvious blood flow signal inside the lesion. While CT scan revealed a cystic lesion (CT value: 20-40 HU) located in the left side of adrenal region, with unclear boundary between itself and neighboring lateral branches of the left adrenal gland (**Figure 1**). Blood pressure, catecholamine, adreno-cortico-tropic-hormone (ACTH)+ cortisol and electrolytes were all in normal range. After sufficient pre-operation preparation, left retroperitoneal cystectomy was performed and patient underwent retroperitoneal laparoscopic surgery under general anesthesia. The adrenal area was fully exposed during the surgery, but the mass was not found. After CT images were reviewed, continual exposure to the abdominal side was conducted until a gourd shape cyst with milk-white content inside (after dissection) could be observed. GD was considered after consultation with gastrointestinal surgeons during the surgery, and the diagnosis was confirmed by endoscopic examination. Gastric diverticectomy was performed and the pathological diagnosis was a xanthelasma. The patient recovered well and was discharged seven days after surgery. The patient was followed up regularly and underwent an annual gastroscopy and CT examination. At time of writing, the patient had been followed for more than two years and their condition was stable.

This study was conducted in accordance with the declaration of Helsinki. This study was conducted with approval from the Ethics Committee

of North Sichuan Medical College. Written informed consent was obtained from the participant.

Discussion

In the present case of GD which was histopathologically confirmed as xanthelasma, and misdiagnosis as an adrenal cyst was initially determined on the basis of an anechoic mass observed according to the initial CT images. However, during the surgery, adrenal gland area cyst was not found.

Xanthelasma is relatively rare in clinic, and it can be found in the digestive tract, esophagus, stomach, small intestine and colon. The stomach is its most common target organ [7], while the adrenal gland area was rarely reported. Gastric xanthelasma is a relatively scarce endoscopic finding, with prevalence from 0.8% to 7% in different study populations [8, 9]. The etiology and pathogenesis of gastric xanthelasma might be related to age, environment, chronic inflammation such as atrophic gastritis and hyperplastic polyps, HP infection, lipid metabolism disorder, autoimmune mechanism and other factors [10]. Gastric xanthelasma might be a sign of pathological aging of gastric mucosa and an alarming sign of early gastric cancer. For instance, Kokal et al [11] performed gastroscopy on 1400 patients, and found 50 xanthelasma cases, they selected another 50 patients without xanthelasma as the control group, and the results suggested that multifocal atrophic gastritis was more common in xanthelasma patients, especially in older patients.

Gastric xanthelasma adrenal cyst

A Japanese epidemiological study retrospectively investigated the relationship between the pathogenesis of gastric xanthelasma and gastric cancer and their respective pathological features. Multivariate analysis showed that gastric carcinoma and gastric xanthelasma were independently correlated [OR and 95% CI: 6.15 (3.95, 9.70), and $P < 0.05$]. There was a significant relationship between gastric xanthelasma and gastric cancer ($P=0.002$). In our case, the patient was a young woman, with superficial gastritis detected by gastroscopy, which was rare in China.

GD is also a rare disease in clinic [12, 13]. No relevant literature reported that GD appeared in the retroperitoneal adrenal region, and at the same time it was histopathologically diagnosed as xanthelasma. It's recognized that the adrenal glands are adjacent to the peritoneal cavity, and some organs such as duodenum, accessory spleen, colon, gastric diverticulum, renal cyst, pancreatic tail tumors and so on might appear in the retroperitoneal region but with lower incidence, about 0.7% as reported [14]. The exact mechanism underlying GD occurring in the adrenal area is not clear, and the possible reason might be that gastric cardia diverticulum breaks into the retroperitoneum adjacent to renal fascia and then enters into the adrenal area during embryonic development according to the present description on GD in embryology [15].

Gastric xanthelasma is a kind of benign lesion with no obvious clinical manifestations, and GD also lacks typical clinical manifestations. A few patients with GD may feel discomfort under the xiphoid process, and some patients might suffer from dysphagia and reflux in severe situations [16]. The prevalence of GD encountered in our CT series is 0.12%. It's especially rare to find GD at left retroperitoneal area [4]. When it is located at the adrenal area and accompanied with hypertension and palpitation symptoms, it could be easily misdiagnosed as adrenal tumors. A search of Pubmed, Medline, EMBASE, Google Scholar, Cnki, Vip and Wanfang databases was conducted, a total of 11 previous studies with complete general information of patients was identified, in which the GD was a pseudotumor of the adrenal gland, and 10 cases with sex and age data (**Table 1**). All studies were case reports of male patients with GD that simulated masses in the left adrenal

region. Combining the literature review and the patient case study, we summarized our experience to reduce misdiagnosis as follows: first, noncontrast CT scanning results were insufficient due to the lack of continuous dynamic observation of the lesion site. It could easily cause misdiagnosis as typically shown in our case, that is, CT showed that the cystic lesion was completely located in the upper left kidney region, only neighboring to the pancreatic tail and aorta before surgery, but the cystic lesion had a close relationship to the stomach after the dynamic observation of cross-sectional images and the coronary images in three dimensional reconstruction. Moreover, when the lesion had similar CT values as gastric contents, neoplasm derived from the gastrointestinal tract should be suspected. Considering relevant literature where nine patients had GD manifested in the pseudotumor in the adrenal area, seven cases had tiny bubbles inside lesions, and two cases had cyst-density like shadow, those lesions should be suspected to originate from the gastrointestinal tract when the tiny bubbles and liquid gas level occurred in CT images [17-20]. Second, besides CT examinations, the endoscopy and digestive tract contrast imaging should be administered to further confirm whether suspected lesions come from the gastrointestinal tract. If misdiagnosis was made before operation and no lesion was detected when the adrenal gland was entirely exposed during the surgical removal of the lesion in adrenal area at posterior abdominal cavity, the gastroscopy could be given to make further confirmation during operation. In this case, the patient did not receive preoperative barium contrast examination, however, complications were avoided as timely gastrointestinal consultation was conducted and clear diagnosis was made by gastroscopy during surgery.

According to relevant literature, GD and gastric xanthelasma are all benign lesions and deserve no surgery, and GD is supported to have an operation only when there are serious clinical symptoms or digestive tract perforations and hemorrhage. Although some literature reported that the gastric xanthelasma might develop into cancer, it still tends to check gastroscopy regularly and eliminate the related risk factors such as chronic gastritis, HP infection, and so on at present. In this case, we made retroperitoneal laparoscopic gastric diverticulectomy because the patient's diverticulum was rup-

Gastric xanthelasma adrenal cyst

Table 1. Summary of reviewed articles describing cases where gastric diverticula were initially misdiagnosed as adrenal tumors in Chinese and English literature

Author	Public year	Cases (N)	Age (y)	Gender	Assessment/imaging finding	Published Journal
Schwartz et al	1986	1	65	M	CT: an incidental 3 × 3 cm, thin-walled cystic mass with air-fluid level adjacent to left adrenal gland. Upper GI barium study: gastric diverticulum.	AJR Am J Roentgenol
Silverman	1986	1	46	M	CT: a soft tissue mass posterior to gastric fundus. Upper GI barium study: gastric diverticulum extending posteriorly from fundus of stomach.	J Comput Assist Tomogr
Chasse et al	2002	1	42	M	CT: a 4.5 cm necrotic mass close to left adrenal gland on upper abdominal. Upper GI barium study: gastric diverticulum.	Surgery
Araki et al	2006	1	47	M	CT: two left adrenal masses; a 3.0 cm lesion in the upper portion of the adrenal and a 1.5 cm diameter lesion in the lower portion.	Int J Urol
Jing and Huang	2007	1	67	M	CT: a 3.4 cm necrotic mass close to left adrenal gland on upper abdominal. Upper GI barium study: a 3 cm gastric diverticulum extending posteriorly from fundus of stomach.	Zhongguo Xian Dai Yi Xue Za Zhi (Chinese)
Kodera et al	2007	1	46	M	CT: a 2.5 cm tumor located in left adrenal region. MRI: tumor clearly isolated from other issues in suprarenal region and exhibited high-intensity area on T2-weighted image. Upper GI barium study: gastric diverticulum.	Endocr J
Nogurea et al	2009	1	N/A	N/A	CT: Small bubble of gas in ventral area of diverticulum. MRI: A small signal void in ventral area interpreted as ferritin and hemosiderin.	Urology
Jebnsingh et al	2014	1	63	M	CT: a well-defined low-density left supra-renal lesion with air fluid level.	BMJ Case Rep
Wang et al	2015	1	15	M	CT: a 2.4 × 1.5 cm cystic mass with air-fluid level. Upper GI barium study: gastric diverticulum.	J Clin Urology (Chinese)
Mahafza et al	2015	1	48	M	MRI: a left adrenal mass. CT with intravenous (IV) contrast media and negative oral contrast media (water): 4 × 4.5 cm non enhancing thin-walled cystic lesion with an air-fluid level in close contact to the gastric fundus posteriorly.	Indian J Surg
Feng	2015	1	49	M	CT: a low-density mass measuring 2.3 cm in diameter in the area of the left adrenal gland.	Oncol Lett

N/A, not available; y, years; N, numbers; M, male; MRI, magnetic resonance image; CT, computed tomography; GI, gastrointestinal.

tured during operational separation, which might cause infection by secretion and the possible diagnosis of a tumor could not be ruled out.

In conclusion, it's rare to find a similar case of GD manifesting as adrenal cyst and was pathologically confirmed as xanthelasma. However, it should be included into the differential diagnosis of retroperitoneal disease to eliminate unnecessary pre-operational examinations such as catecholamine, ACTH and cortisol tests and surgery. The review of this case is for significance of GD and xanthelasma recognition and clinical practice.

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Disclosure of conflict of interest

None.

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